

Measuring and training of walking abilities in pediatric neurorehabilitation

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SUMMARY

Mobility, or the ability to move or be moved in one's environment with ease and without restrictions, is a relevant factor regarding physical independence and social participation. Many children with acquired or congenital neuromotor disorders are faced with limitations concerning their independent mobility, with restrictions in their walking abilities playing a vital role in this respect. Hence, an essential goal of neuropediatric rehabilitation is to influence children's walking abilities positively. This thesis focuses on two different areas that are closely interlinked within rehabilitation: the measurement and the training of functional walking abilities in children with neuromotor disorders.

CHAPTER 1 describes the significance of mobility and how the mobility of children with neuromotor disorders is affected. With the International Classification of Functioning, Disability and Health for Children and Youth (ICF-CY), it introduces an internationally accepted and widely used framework that facilitates a holistic view to document and measure a child's health and disability status and to plan or evaluate interventions in clinical and research settings. Cerebral palsy (CP) - with an incidence rate of 2-3 per 1'000 live births one of the most common congenital disabilities - and its consequences on various health dimensions and possible secondary impairments are discussed. The chapter also addresses the expanded and revised Gross Motor Function Classification System (GMFCS - E&R), which can be considered an essential standard in clinical practice and research to classify the gross motor function of children with CP. It is based on categorizing self-initiated movements with an emphasis on sitting, walking, and wheeled mobility. Furthermore, the chapter gives an insight into the multidisciplinary and the content of pediatric neurorehabilitation and discusses various treatment approaches affecting gait function. The evidence of these different gait interventions and their implementation modes is discussed with a particular focus on robot-assisted gait training (RAGT). Additionally, several aspects that should be considered when measuring walking ability are addressed: The influence of the environment, which is reflected in the different constructs of performance and capacity, and the

psychometric properties of an outcome measure, which should be taken note of depending on the measurement goals. The chapter ends with the outline and the aims of this thesis, addressing the evaluation and the training of gait function. The aims were to:

- (1) investigate and summarize the evidence of gait instruments and their psychometric properties in neuropediatric populations
- (2) gain knowledge of the psychometric properties of selected measures reflecting functional mobility performance in our inpatient neurorehabilitation population
- (3) evaluate the effectiveness of RAGT in children with CP.

CHAPTER 2 systematically investigates and summarizes the evidence of gait capacity and performance measures with the ICF-CY as a reference framework and evaluates their psychometric properties in neuropediatric populations. Our systematic review provided an overview of the current evidence on the psychometric properties of 27 measures used to assess functional gait in children with neuromotor disorders. CP was the diagnosis most often considered in studies examining the psychometric properties of a measure. Most studies evaluated aspects of reliability. Studies on measurement error and especially responsiveness were the rarest. The studies' sample sizes were generally low. Our review revealed a substantial lack of evidence regarding the psychometric properties of many measures that are regularly used in research and clinical practice. Furthermore, only a few of the appraised measures addressed the performance level of the ICF-CY, even though these measures take the influence of the environment children are exposed to in real life into account, which we consider very relevant.

Based on the findings of the systematic review and the underrepresentation of performance measures in our neurorehabilitation inpatient setting, we considered the Functional Mobility Scale (FMS) and the Functional Assessment Questionnaire – walking scale (FAQ) to be the most suitable tools to be applied in our setting, regarding their practicability and usability. Both are performance measures that address functional mobility in the daily environment. To be used in our rehabilitation center, we translated the FMS and FAQ into German and evaluated their

psychometric properties in children and adolescents with neuromotor disorders, who were inpatients in our center.

CHAPTER 3 reports on the translation and the examined concurrent validity of these two measures, by comparing them to well-known activity measures assessing walking abilities in children, such as the walking item of the Functional Independence Measure for Children (WeeFIM walking), the 10-meter (10MWT) and 6-minute walk tests (6MinWT), and the dimension E (walking, running, jumping) of the Gross Motor Function Measure-88 (GMFM E). All correlation coefficients were moderate to strong, exceeded the hypothesized lower limits, and corresponded well with the reported values of the original English versions. However, correlations with the WeeFIM walking, which is also a performance measure, were not higher than correlations with the capacity measures. A possible explanation for this finding could be that the environment in the rehabilitation center with the optimized infrastructure and the daily routine of the inpatient setting may have reduced the differences between walking performance and walking capacity.

Both measures did not seem to be affected by any floor or ceiling effects in our patient sample, and none of the correlation coefficients exceeded 0.8. Thus, the FMS and FAQ provided additional information on functional mobility in the inpatient setting and facilitated a more differentiated picture of patients' mobility levels in varying contexts during the inpatient stay. Measuring performance hereby provides a meaningful perspective for the child and their family as the results are directly transferable to the child's behavior in everyday life.

Our study expands the already existing evidence of the FMS and FAQ's validity to assess functional mobility also in the inpatient setting.

CHAPTER 4 determines the interrater reliability of the FMS and FAQ. We investigated several sources of variation regarding the raters and the environment by comparing the ratings between nurses and physiotherapists in our inpatient neurorehabilitation setting and between these health professionals at one side and the parents' ratings in the home environment on the other side. We performed these comparisons twice; at the start and end of active gait rehabilitation. By choosing such a design - with different characteristics between the groups of raters, different

settings, and two time points to investigate - we wanted to examine whether the scores of children's performance levels of functional mobility are comparable between a home and an inpatient setting despite differences in the environment and the raters' background. Furthermore, we wanted to know whether the timespan a health professional has worked with an individual child influences the rating.

With weighted kappa values of 0.69-0.77 for the FAQ and 0.62-0.92 for the FMS, interrater agreements between physiotherapists, nurses, and parents were above our predefined acceptance level of 0.6 at both time points. Our results indicate that functional mobility performance in the inpatient setting and the children's everyday environment seem to correspond well with each other and that all rater groups score the FMS and FAQ equally.

CHAPTER 5 explores the responsiveness and minimally important change (MIC) of the FMS and FAQ. For this purpose, we applied a construct-based approach (i.e., we examined the correlations between changes over time in FMS/FAQ scores to changes over time in standard functional gait measures) and a criterion-based approach (i.e., we introduced a 5-point ordinal global rating scale (GRS) to quantify the physiotherapists' perceived change of children's functional mobility performance after inpatient rehabilitation). Both methods are recommended by the CONsensus-based Standards for the selection of health Measurement INstruments (COSMIN) group. In line with these recommendations, we defined a priori hypotheses about the expected results. Regarding the construct approach, change scores of the FMS/FAQ correlated between 0.54-0.76 with the WeeFIM walking, the other performance measure, 0.35-0.49 with capacity measures like the 10MWT, 6MinWT, and GMFME, and 0.57-0.76 with the GRS. We dichotomized the GRS with the physiotherapists' perceived change of functional mobility performance (unchanged and improved) and used it as a criterion to perform receiver operating characteristic (ROC) curves analyses. The values of the area under the ROC curves (AUC), which were derived from this criterion approach, were above the preset acceptance level of 0.7. Exceeding this limit demonstrated the measures' ability to accurately discriminate between children who did or did not improve according to the therapists' appraisal.

Using the GRS, we further obtained a definition of what therapists consider as minimally important change. The MIC values indicated a 1-point increase for the FMS and a 2-point increase for the FAQ to be clinically meaningful.

In summary, we have demonstrated in chapters 3 to 5 that the FMS and FAQ are valid and reliable tools to assess the functional mobility performance of children with neuromotor disorders not only in their everyday home environment but also in the inpatient rehabilitation setting. Further, these measures can be used to detect changes in functional mobility performance that have occurred during inpatient rehabilitation, and thresholds were determined which define whether an observed change can be considered clinically meaningful.

CHAPTER 6 describes the study protocol and presents the theoretical considerations of a pragmatic crossover trial on the effectiveness of RAGT on the Lokomat compared to usual care. Children aged 6-18 years with bilateral spastic CP who could walk at least 14 meters with or without walking device were recruited. They were randomized to two different pre-specified sequences of interventions. One intervention was RAGT as treatment (T) on the Lokomat in the outpatient setting of our rehabilitation center, consisting of three weekly sessions over five weeks. The other intervention was usual care as control (C) in the home environment of the child (consisting of 1-2 sessions of physiotherapy per week and additional hippotherapy, circuit training, and occupational therapy as necessary). A child could be randomized to a T/C sequence (TC-group) or a C/T/C sequence (CTC-group). Outcome measures included the GMFM E as the primary outcome, and the dimension D (standing) of the GMFM-88 (GMFM D), as well as 6MinWT and 10MWT as secondary outcomes – assessed before and at the end of each intervention period. A sample size calculation indicated a sample size of 30 to be sufficient to detect a difference of 3.7%-points in the dimension E-score of the GMFM-88. We increased this number to 34 to allow for a dropout rate of about 10% and planned a trial duration of four years.

CHAPTER 7 presents the findings of the pragmatic, randomized, crossover trial described in chapter 6 and reflects on the lessons we learned from conducting this trial. We stopped the study prematurely after more than eight years and after 16 of

the planned 30 patients had finished the protocol. Our results indicated that five weeks of RAGT, applied as a standalone therapy intervention, were not superior to five weeks of usual care regarding the improvement of walking abilities in ambulatory children with spastic CP. Although these results may have been due to the underpowered study, the effect sizes of the primary, as well as the secondary outcome measures (except for the 6MinWT, which showed a small effect size), do not suggest any other conclusion.

Performing this trial, designed more than nine years ago as explained in chapter 6, has taught us a lot from a content-related as well as methodological point of view. We had aimed for a pragmatic trial design. However, the choices we made regarding the setting and recruitment turned out to be more on the explanatory side, which emerged as aggravating factors in the course of the study. Although we also provide outpatient therapy in our clinic, the RAGT intervention on the outpatient basis proved to be the main reason for our recruitment problems. Most families preferred to arrange the RAGT period during their child's school holidays, to limit the burden on their child due to therapy and travel expenses as much as possible. This fact reduced the number of participants considerably. Contrary to earlier conventions of 10 years ago, when we had planned our study, current clinical recommendations are to embed RAGT as an add-on therapy element within an individualized holistic treatment package. This approach also reflects the common practice of today's rehabilitation and thus contributes to producing research results that are relevant and applicable in practice. Considering performance in everyday life and addressing patients' personal goals when applying outcome measures are further factors to increase the relevance of research results for families and health professionals.

CHAPTER 8 summarizes the main findings, discusses methodological considerations, addresses implications for future research and clinical practice, and concludes with the main contributions of this thesis. We highlighted the lacking evidence on psychometric properties of measures assessing walking abilities in children with neuromotor disorders and the general shortage of performance measures regarding children's functional mobility in their daily environment. This thesis reduced the gap regarding both of these issues by providing evidence for

aspects of validity, reliability, and responsiveness of the German versions of the FMS and FAQ in the inpatient setting. Further, we facilitated the interpretation of change scores by defining MIC values for both measures.

As the crossover trial indicated that RAGT on an outpatient basis was not effective in improving walking abilities in ambulatory children with CP, evidence on the effectiveness of pediatric RAGT is still vague. Because of the small sample sizes, heterogeneous patient populations, and special needs, intervention studies in the pediatric field remain challenging and ask for innovative research approaches.